

Rare Cause of Pleural Nodularity: Splenosis

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Splenosis is a rare condition described as ectopic splenic tissue implantation generally after a splenic rupture.

A 35-year-old male patient who had a history of splenectomy operation due to gunshot wound seven years ago was referred to our hospital with complaints, including exhaustion, sweating and shortness of breath. Thoracic computed tomography of the patient showed nodular pleural thickenings at the level of the left lower lobe, which proved to be unrelated with the presenting symptoms. The patient underwent a minithoracotomy for diagnosis and treatment. During the intraoperative observation, dark-colored, soft, multiple nodular lesions with a biggest size of 2 cm inside the visceral pleura over an area of 5 x 10 cm in dimension were observed. Also, a few tiny nodules in the lung parenchyma approximately 1-cm deep to the pleural nodules were palpated. The lesions were excised. The histopathological examination of the specimen showed splenic tissue in the lung parenchyma and pleura, so the lesions were accepted as splenosis.

Pleuropulmonary splenosis, which develops generally after simultaneous rupture of the diaphragm and spleen, is a very rare condition. Most of the patients are asymptomatic and the lesions are detected accidentally. If the diagnosis can be made preoperatively, surgical excision is not needed.

Key words: pleural nodularity ■ pleuropulmonary splenosis ■ spleen

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INTRODUCTION

Splenosis is a rare condition described as ectopic splenic tissue implantation generally after a splenic rupture. Most of the reported cases are intraabdominal, and thoracic splenosis is even much rarer.¹ The lesions generally look as asymptomatic pleural-based nodules, and noninvasive diagnosis requires high index of suspicion.² In this report, we present a case of thoracic splenosis, which is either pulmonary parenchy-

mal and pleural. The patient was diagnosed seven years after an abdominal gunshot wound.

CASE REPORT

A 35-year-old male patient who had a history of splenectomy operation due to gunshot wound seven years ago was referred to our hospital with complaints, including exhaustion, sweating and shortness of breath. Physical examination was normal except a laparotomy scar. Chest x-ray did not reveal any pathology except a minimal blunting of left costophrenic angle. Thoracic computed tomography (CT) of the patient showed nodular pleural thickenings at the level of the left lower lobe and over the left hemidiaphragm (Figure 1). A CT-guided Tru-Cut needle biopsy was nondiagnostic. Also video-assisted thoracoscopic surgery was not considered because of possible pleural adhesions, so the patient underwent a minithoracotomy for diagnosis and treatment. During the intraoperative observation, dark-colored, soft, multiple nodular lesions with a biggest size of 2 cm inside the visceral pleura over an area of 5 x 10 cm in dimension were observed. Also, a few tiny nodules in the lung parenchyma approximately 1-cm deep to the pleural nodules were palpated. After observing color and softness of the lesions and combining this with the history of the patient's splenic trauma, we suspected an ectopic splenic tissue. The lesions were excised by pleurectomy and wedge resection of the lung. The histopathological examination of the specimen showed splenic tissue in the lung parenchyma and pleura, so the lesions were accepted as splenosis (Figure 2). After the diagnosis of splenosis, an abdominal CT was performed and a 2 x 1-cm mass was detected in the left retrocrural area of the upper abdomen. A technetium-99m sulphur colloid radionuclide scan confirmed the abdominal splenosis. The patient's complaints did not appear to be directly due to splenosis, and any specific underlying etiological factor could not be found. Therefore, the patient is still under follow-up.

DISCUSSION

Splenosis represents the autotransplantation and regrowth of splenic tissue after splenic trauma or surgery. It has to be distinguished from accessory spleens, which are congenital and characterized by a mainly left

abdominal location and from so-called splenic-anlage tumors, which are rare hamartomatous splenic heterotopias in the liver.³

Most of the splenosis cases involve the abdomen. Rarely, splenic tissue may gain access into the thorax either from a diaphragmatic tear or through diaphragmatic foramina, and seed the pleural space.⁴ Splenic implants derive their blood supply from surrounding tissue and grow into mature splenic tissue.⁵ They present themselves usually as pleural-based, extraparenchymal nodules in the left hemithorax but rarely settle into the pulmonary parenchyma through a concomitant parenchymal laceration.^{1,4} Normand and coworkers found abdominal or pelvic splenosis and thoracic splenosis in 65% and 18% of concomitant splenic and diaphragmatic injury cases, respectively.⁶ We found 45 thoracic splenosis and seven pulmonary parenchymal splenosis cases reported in the English literature.^{1,6,7} In our patient, we could not see any finding of diaphragmatic rupture during surgery. Splenic implants might have passed through a diaphragmatic hiatus or a very

small diaphragmatic rupture. Also, there must have been a lung parenchyma laceration, resulting in pulmonary parenchymal splenosis.

Although a vast majority of the patients with thoracic splenosis are asymptomatic, a few cases with hemoptysis and pleurisy were reported.^{8,9} Cordier and coworkers attributed their patients' hemoptysis to thoracic splenosis because they could not find any other potentially causative thoracic disease. Furthermore, the hemoptysis had clearly improved after resection of the splenotic lesion.⁹ Also, Singh and coworkers reported a case with pleuritic pain but did not mention about the mechanism of pleurisy.¹⁰

Radiological studies such as chest x-ray or thoracic CT are nonspecific and show the lesions as pleural-based pulmonary nodules. Differential diagnosis includes pleural metastases, lymphoma, asbestos-related pleural plaques, mesothelioma and invasive thymoma with contiguous extension.⁸ Although fine-needle aspiration is most often inconclusive, Tru-Cut (Allergiance Healthcare Corp., McGaw Park, IL) needle biopsy has provided enough tissue for histologic diagnosis in some reports.¹¹ The absence of Howell-Jolly bodies, pit-

Figure 1. Thoracic computed tomography: nodular pleural thickenings at the level of left lower lobe and over the left hemidiaphragm

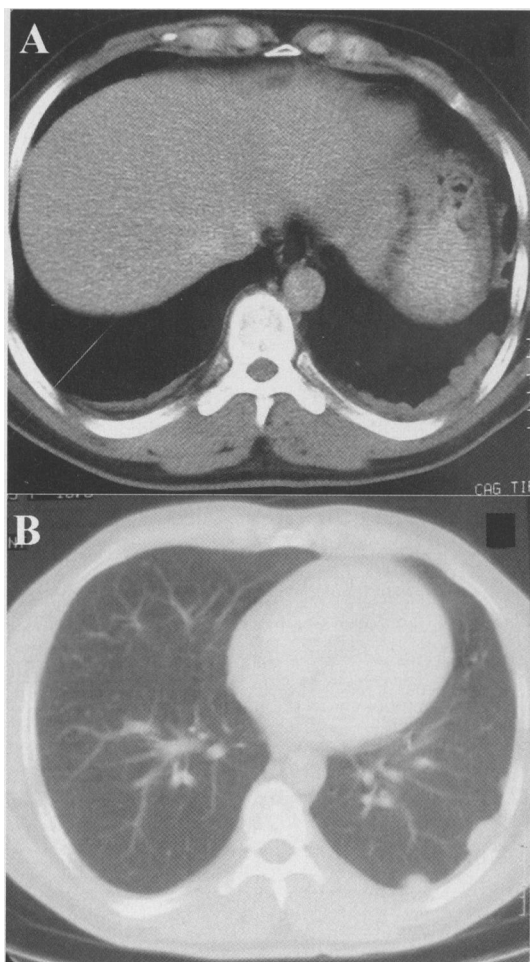
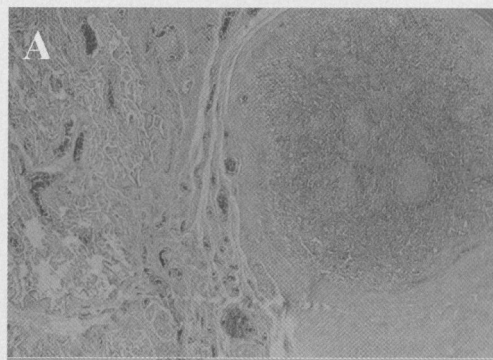
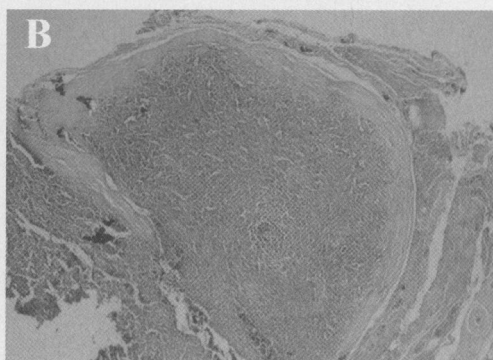


Figure 2. Microscopy of pulmonary splenosis

A. Splenic tissue characterized with white and red pulpa surrounded by a thin fibrous capsule in the lung parenchyma (H&E x40)



B. White pulpa presenting artery in the center and red pulpa characterized with sinusoids (H&E x40)



ted erythrocytes and siderocytes in the peripheral blood in patients who have had splenectomy suggests the presence of residual splenic tissue. The diagnosis of thoracic splenosis can be established preoperatively with radionuclide scans using technetium-99m sulfur colloid, indium-111-labeled platelets and technetium-99m heat-damaged erythrocytes (sequestered in the reticuloendothelial system and thus in any ectopic splenic tissue).^{5,6} Although nuclear medicine is the mainstay in the diagnosis of splenosis, magnetic resonance imaging (MRI) with superparamagnetic iron oxide combines a physiologic test of reticuloendothelial cell uptake with the anatomical details of MRI. This may be helpful in detecting splenic tissues.¹² In our patient, the diagnosis of thoracic splenosis was not suspected before the surgery as were most of the other reported cases. Therefore, a study of radionuclide scan or superparamagnetic iron-oxide-enhanced MRI was not considered before the surgery.

If preoperative diagnosis can be established, resection of splenotic lesions is unnecessary. Even, these implanted splenic tissues offer some degree of protection against bacterial infection, lowering the frequency of postsplenectomy sepsis in an asplenic patient.⁷ Hatway and coworkers found normal immunological status in an asplenic patient who had splenotic nodules.¹³ Also, Pearson and coworkers found more frequent return of splenic function and lower rates of bacterial sepsis in asplenic patients with splenosis.¹⁴ Therefore, we left our patient's retrocrural splenotic nodule in place.

In conclusion, a high index of suspicion is needed for the preoperative diagnosis of thoracic splenosis. By using radionuclide scans or superparamagnetic iron-oxide-enhanced MRI in left-sided pleuropulmonary-nodule patients with a history of thoracoabdominal trauma, some of the unnecessary biopsies can be avoided.

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